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Determinants of health-related and oral health-related quality of life in adults with orofacial clefts: a cross-sectional study

ABSTRACT

Objectives: To investigate the structural and intermediary determinants of health-related quality of life (HRQoL) and oral health-related quality of life (OHRQoL) among adults with cleft lip and/or palate (CL/P).

Design and Participants: A cross-sectional study was conducted with patients enrolled at the referral center for craniofacial anomalies in Manaus, Brazil. Adults aged 18 years or more with non-syndromic CL/P were selected.

Main Outcome Measures: HRQoL and OHRQoL were assessed using the 36-Item Short Form Health Survey (SF-36) and the Oral Impacts on Daily Performance (OIDP), respectively. Individual interviews and oral examinations were conducted to collect data on socioeconomic and demographic characteristics, social ties, health-related behaviors, compliance of CL/P protocol, chronic diseases, type of CL/P, oral clinical measures and CL/P related measures. Poisson regression was used to test the association of independent variables with HRQoL and OHRQoL outcomes.

Results: The mean age of the 96 participants was 29.4 ± 9.1 years. Low family income, female sex, low social support, type of CL/P and dental caries were associated with poor HQoL and poor OHRQoL ($P < .05$). Poor HRQoL was also associated with chronic diseases ($P < .05$). Adults with low education, low social network and smokers were more likely to have worse OHRQoL ($P < .05$).

Conclusions: Structural and intermediary determinants were related to HRQoL and OHRQoL in adults with CL/P suggesting the need for interdisciplinary approaches to improve the management of CL/P and intersectoral actions to reduce the impact of social inequalities.

Keywords: quality of life, cleft lip, cleft palate, adults

The treatment needs of individuals with cleft lip and/or palate (CL/P) has been predominantly evaluated through clinical measures (Brattström et al., 2005; Mølsted et al., 2005). However, the biomedical model has been replaced by the socioenvironmental model of health emphasizing the importance of social environments as major determinants of health leading to a paradigm shift in health care (World Health Organization, 2010). In addition, the development of patient-reported outcome measures, such as quality of life (QoL) measures, has contributed to a better understanding of the effectiveness of the treatment of individuals with CL/P (Nettleton, 1995).

Facial disfigurement and functional impairment as a result of craniofacial conditions can negatively impact on individuals' QoL and well-being (Rumsey and Harcourt, 2004). Although patients with craniofacial anomalies can live without major psychological problems, individuals with CL/P are more prone to psychological impairments, including social isolation and low self-esteem (Turner et al., 1998; Endriga and Kapp-Simon, 1999; Hunt et al., 2005). Patients with CL/P may also have long-term psychosocial problems even after they have completed the treatment (Hunt et al., 2005). In addition to psychological and social disorders, physical impairments related to aesthetics and functional handicap influence the QoL of individuals with CL/P (Mani et al., 2010; Foo et al., 2012; Herkrath et al., 2015).

Quality of life can be defined as "individuals' perceptions of their position in life in the context of the culture and value systems in which they live. It also refers to their goals, expectations, standards, and concerns" (World Health Organization Quality of Life Group, 1997). Health-related quality of life (HRQoL) and Oral health-related quality of life (OHRQoL) are components of QoL. HRQoL is a multidimensional construct related to the perceived impact of health status on the different aspects of the everyday life (Fleck et al., 1999). OHRQoL refers to the individuals' perception of the symptoms and functional and psychosocial impacts affected by oral disorders (Locker and Allen, 2007).

There is no consensus on the influence of orofacial clefts on HRQoL and OHRQoL. Although HRQoL measures do not differ between adults with and without CL/P in some studies, other studies report that individuals with CL/P experience poorer QoL compared to those without this condition (Marcusson et al., 2001; Sinko et al., 2005; Oosterkamp et al., 2007). In a recent meta-analysis, adults with CL/P reported worse HRQoL than those without oral clefts (Herkrath et al., 2015). There is no agreement on the possible impact of CL/P on OHRQoL in adults, indicating that such a relationship remains unclear (Munz et al., 2011; Foo et al., 2012).

Despite the increase in the number of studies involving QoL measures, little attention has been paid to factors associated with HRQoL and OHRQoL in individuals with CL/P. Previous studies on HRQoL and OHRQoL in patients with CL/P have focused on the characteristics of treatment and satisfaction with facial appearance and function. However, these characteristics may explain only part of the variation of the QoL measures. In the general population, socioeconomic and demographic characteristics, social ties, health-related behaviors, clinical conditions, self-perceived general health have been associated with HRQoL (Michelson et al., 2001; Mitra et al., 2004; Acree et al., 2006; Lua et al., 2007; Al-Naggar et al., 2011; Brennan and Spencer, 2012; Kumar et al., 2014). Most studies on the potential determinants of HRQoL and OHRQoL in patients with CL/P have been limited to socioeconomic and demographic factors (Mani et al., 2010, Broder et al., 2012; Dak-Albab and Dashash, 2013; Broder et al., 2014b). In addition, psychosocial factors, including depression and self-efficacy, and surgical treatment needs have been found to predict OHRQoL in young people (Broder et al. 2014a). As far as we know, no previous study evaluated the determinants of HRQoL and OHRQoL in adults with CL/P using a theoretical model.

A conceptual framework on the determinants of health inequalities was proposed by the World Health Organization (WHO) Commission on the Social Determinants of Health

(CSDH). The WHO framework encompasses structural determinants (e.g. demographic factors and socioeconomic position) and intermediary determinants (eg. psychosocial factors, behaviors, biological factors and health services) that influence health outcomes (World Health Organization, 2010). The present study adopted the WHO theoretical model to investigate the determinants of HRQoL and OHRQoL in adults with non-syndromic CL/P (Figure 1). The aim of this study was to test the relationship of structural (socioeconomic and demographic factors) and intermediary determinants (social ties, behaviors and biological factors) with HRQoL and OHRQoL in adults with non-syndromic CL/P. The present study was conducted in the North of Brazil, which is considered the most socially deprived region of the country with strong inequalities in health and healthcare provision. Thus, the identification of associated factors of HRQoL and OHRQoL in individuals with CL/P in this region can contribute to the development of local strategies and the identification of needed interventions to improve their quality of life and well-being.

METHODS

Study design and participants

A cross-sectional study was conducted with adults with CL/P enrolled at the referral center for patients with congenital malformations in the city of Manaus, Brazil. The center was the only public service for the management of individuals with CL/P in the State of Amazonas, Brazil. All newborns with CL/P in the State of Amazonas are referred to the referral center by the neonatologists or pediatricians for enrollment using a referral form. Parents of newborns with CL/P receive information about the types of treatment provided by the referral center and the contact details before children's maternity discharge. Nealy 95% of the individuals with CL/P living in the State of Amazonas are registered at the referral center. Individuals with CL/P living in the city of Manaus who were not born in the State of

Amazonas are enrolled at the referral center through the reference system administered by the local health care system. There are few other private health services that offer specific medical/dental treatments for the management of individuals with CL/P in the state. These services do not provide comprehensive care for individuals with CL/P, do not offer complex procedures and do not operate in a referral health care model.

The studied population included subjects aged 18 years or older living in Manaus who had completed the repairing treatment for oral clefts or those without access to additional treatment. The latter criterion was based on the capacity of the local referral center to provide some types of complex treatments for patients with oral clefts. Thus, patients with CL/P with recommendations of additional treatments for oral clefts but without access to them were also included in the study. Individuals with syndromes, other congenital anomalies or acquired craniofacial disfigurement were excluded.

Procedures

A list of all persons with oral clefts registered at the referral center for patients with congenital malformations in 2013 Manaus was obtained. Adults aged 18 years or more were initially contacted by telephone. Home visits were conducted to invite people who could not be reached by telephone to participate. Since it was anticipated that some cases were not registered at the referral center, all participants were asked to inform about other people with CL/P they might know as well as their contact details. They were invited to participate after receiving the information about the objectives of the study during the initial contact. After signing an informed consent form, participants were interviewed and examined.

Primary data were collected through in-person interviews between January and September 2013. A trained interviewer conducted individual interviews using standardized questionnaires and a calibrated dentist carried out clinical oral examinations in the dental office.

The study was approved by the Committee of Ethics and Research of the National School of Public Health, Oswaldo Cruz Foundation (protocol CAAE n° 06261412.7.3001.5441).

Measures

Health outcomes

Health-related quality of life

The validated version of the Medical Outcomes Study SF-36 item short-form survey questionnaire (SF-36) for Brazilian adults was used to evaluate HRQoL (Ware and Sherbourne, 1992; Ciconelli et al., 1999). The SF-36 is composed of 36 items aggregated into 8 domains as follows, ‘physical functioning’, ‘role limitations due to physical health’, ‘role limitations due to emotional problems’, ‘vitality’, ‘mental health’, ‘social functioning’, ‘bodily pain’ and ‘general health’. The responses of the items are converted into a centesimal scale to generate the domain scores which are the average of the related items. The overall score is the average of the domain scores, ranging from 0 to 100. The higher the score, the better the HRQoL.

Oral health-related quality of life

OHRQoL was evaluated using Oral Impacts on Daily Performance (OIDP) questionnaire validated for the Brazilian population (Adulyanon and Sheiham, 1996; Cortes et al., 2002). The OIDP measures the oral impacts on the person’s ability to perform daily activities in the last six months considering eight performances as follows, ‘eating’, ‘speaking’, ‘cleaning teeth’, ‘sleeping and relaxing’, ‘smiling and showing teeth’, ‘emotional status’, ‘carrying out major work or role’ and ‘social contact’. This questionnaire involves asking participants to report on daily performances that have been impacted by their oral health status. They then describe the frequency of these impacts, ranging from 0 (never affected in the past 6 months) to 5 (being affected every day or nearly every day). They are also asked to rate the intensity of the impacts, ranging from 0 (none) to 5 (very severe). The

score in each performance is calculated by multiplying the frequency with the intensity score. The overall OIDP score is the sum of the scores of the eight performances with a maximum value of 200 (8 x 5 x 5). A higher OIDP score indicates worse OHRQoL.

Structural determinants

Socioeconomic position

Socioeconomic position included education, family income, and marital status. Education was assessed based on years of schooling. Family income was recorded according to the Brazilian minimum wages (BMW) (R\$678.00, corresponding to \$339.00) and then categorized as ≤ 2 and > 2 BMW. Marital status was recorded as single, married or living with a partner, separated or divorced and widowed.

Demographic factors

Demographic data were age and sex.

Intermediary determinants

Social ties

Social ties were assessed through social support scale (Sherbourne and Stewart, 1991) and a social network measure of friends proposed by Berkman and Syme (1979) adapted for the Brazilian population. Cronbach's alpha for social support scale was 0.94. The questionnaires showed adequate psychometric properties (Chor et al., 2001). The social support questionnaire consisted of 19 items comprising five domains: 'material support', 'affectionate support', 'emotional support', 'positive social interaction' and 'informational support' (Sherbourne and Stewart, 1991). The total score was calculated through using the average of the dimension scores, converted into a centesimal scale. Social networks were assessed using a question concerning the person's relationship with their friends. Participants were asked to state how many friends they feel comfortable with and who they can talk to about almost everything (Berkman and Syme, 1979).

Health-related behaviors

Health-related behaviors included current smoking (yes, no), risk of alcoholism assessed using the CAGE questionnaire (Masur and Monteiro, 1983) and physical activity in the last 7 days (none and ≥ 1 days).

Biological factors

Biological measures included compliance with the treatment protocol for patients CL/P, chronic diseases, type of oral cleft and oral clinical measures. Compliance with the protocol of CL/P treatment was evaluated according to the guidelines proposed by the Rehabilitation Hospital for Craniofacial Anomalies at the University of São Paulo (Freitas et al., 2012). Patients were grouped as follows: ‘not submitted to primary plastic surgery’; ‘only primary plastic surgery’; and ‘additional procedures after the primary plastic surgeries’.

Each participant was asked whether they had one or more of the following chronic diseases: diabetes, hypertension, joint disease, cancer and respiratory disease. These diseases were selected because they were considered the most prevalent chronic conditions in adults by the World Health Organization (World Health Organization, 2011).

Clinical oral examinations were performed by a single examiner to assess the types of oral clefts as follows: cleft lip with or without cleft alveolus (CL \pm A), cleft palate (CP) and cleft lip and palate (CLP). Plan dental mirrors and WHO probes (Golgran ®) were used to register dental caries (DMFT index), number of decayed teeth, number of missing teeth, and the malocclusion (Dental Aesthetic Index/DAI) of participants (World Health Organization, 1997).

CL/P related measures

CL/P related measures were registered to describe the main characteristics of participants with different types of CL/P. They included treatment history of CL/P, craniofacial growth, aesthetic appearance of the nasolabial region, satisfaction with facial appearance, function, and CL/P treatment. CL/P related measures were not considered in the

statistical modeling due to collinearity and missing data. Some measures were registered only for some oral cleft types and this led to some missing data.

Treatment history of CL/P

Treatment history of CL/P included the age of the participant when the first lip repair and palatoplasty was performed. Self-reported information included whether the participant had other procedures related to CL/P care including; orthodontics, orthognathic surgery, speech therapy, and other plastic surgeries.

Craniofacial growth

The interarch occlusal relationship of patients with complete unilateral and bilateral cleft lip and palate (UCLP and BCLP) was evaluated using the Goslon yardstick and the Bauru index (BCLP yardstick), respectively, as a proxy measure of craniofacial growth. The measures assess anteroposterior, transverse and vertical interarch discrepancies (Mølsted et al., 2005).

Aesthetic appearance of nasolabial region

The nasolabial aesthetics of participants was evaluated according to the score proposed by Asher-McDade et al. (1991) and the Eurocleft and Americleft intercentre collaborative studies (Brattström et al., 2005; Mercado et al., 2011). The nasolabial aesthetics measure considers four components: (i) nasolabial morphology (nasal form); (ii) nasal symmetry (deviation of the nose); (iii) redness border of upper lip (shape of vermilion border); and (iv) nasal profile including upper lip. Each component is assessed using a 5-point scale, ranging from 1 (very good appearance) to 5 (very poor appearance) (Kujipers-Jagtman et al., 2009). The total score is obtained from the average of the four components. This measure was originally developed for individuals with unilateral cleft lip and palate (UCLP). Participants with unilateral CL±A were also assessed since this cleft type imposes aesthetic impacts on similar areas.

Satisfaction with facial appearance and function and satisfaction with CL/P treatment

The Cleft Evaluation Profile (CEP) instrument was used to assess patient satisfaction according to eight CL/P-related features (speech, hearing, teeth, lip, nose, nasal breathing, facial profile, occlusion) through a 7-point Likert scale (Turner et al., 1997), in which 1 represented completely dissatisfied and 7 completely satisfied with the feature evaluated. In addition, the overall satisfaction with CL/P treatment was assessed on a 5-point Likert scale, ranging from very satisfied (1) to very unsatisfied (5) (Roncalli et al., 2012). The greater the score the worse the overall satisfaction.

Pilot study and clinical calibration

Ten subjects aged 18 years or more with CL/P under treatment were selected by convenience sampling in the same referral center for patients with congenital malformations. They were examined and interviewed twice at a seven-day interval. The intra-examiner intraclass correlation coefficient (ICC) for DAI was 0.813. The Kappa coefficients for DMFT index, Goslon/Bauru yardstick and the nasolabial region aesthetic components ranged from 0.786 to 1.000. ICC of agreement for social support scale and for social network of friends were 0.94 and 0.89, respectively.

Quality assurance

The quality of the data from the main study was assessed through the replication of interviews and clinical exams in 10% of the sample at an interval of seven days. One in every ten individuals was selected and reassessed. ICC of HRQoL and OHRQoL measures were 0.693 and 0.873, respectively. The ICC of clinical oral measures were 0.715 for nasal morphology score, 0.736 for upper lip vermilion score, 0.761 for DAI, 0.767 for the nasal deviation, and 1.000 for the DMFT index, Goslon/Bauru yardstick index and nasal profile.

Statistical analysis

Socioeconomic factors, demographic characteristics, social ties, health behaviors, biological measures, chronic diseases, oral clinical measures, treatment history of CL/P, craniofacial growth, aesthetic appearance of nasolabial region, satisfaction with facial

appearance and function and satisfaction with CL/P treatment were described for the whole sample and according to the CL/P groups: CL±A, CP and CLP through means (standard deviation), and proportions. The mean and standard deviation of the total score and item scores of OIDP questionnaire and the overall score and the domain scores of the SF-36 questionnaire were presented for the whole sample and according to the CL/P groups. The median and range of OIDP and SF-36 total scores were also reported. The comparison of OIDP and SF-36 scores between CL/P groups was assessed using Kruskal-Wallis.

Psychometric properties of the OIDP and SF-36 questionnaires were assessed through internal consistency using Cronbach's α coefficient. Correlation coefficients between OIDP items scores and OIDP total score and SF-36 domains scores and SF-36 total score were described.

The normal distribution of OIDP scores and SF-36 scores was tested using the Kolmogorov-Smirnov test. Both QoL outcome measures demonstrated a skewed distribution and the hypothesis of normality of OIDP and SF-36 scores were rejected ($P < .001$). Consequently, Poisson regression was used to assess the relationship of structural and intermediary determinants with SF-36 and OIDP total scores using robust variance to account for data overdispersion. Coefficients and standard errors (SE) of the unadjusted associations between each independent variable and QoL measures were initially obtained. Variables that were significant at 5% ($P < .05$) were used in the multivariate analysis.

Multivariate Poisson regression using nested models were used to test the association of structural and intermediary determinants with SF-36 and OIDP scores. Sequential modeling using the stepwise forward procedure to select independent variables was carried out to test five models according to the theoretical model presented in Figure 1. The independent variables were grouped according to the structural and intermediary determinants of HRQoL and OHRQoL (World Health Organization, 2010). Structural determinants included socioeconomic factors (family income, education, and marital status)

and demographic characteristics (age and sex). Intermediary determinants were composed by social ties (social support and social network), health-related behaviors (smoking, risk of alcoholism, physical activity) and biological factors (CL/P treatment protocol compliance, chronic disease, type of oral cleft and oral clinical measures) (Figure 1). The association between socioeconomic factors and QoL measures was tested in Model 1. Demographic data were added to Model 2, sequentially social ties in Model 3, health behaviors in Model 4 and biological measures in Model 5. Independent variables of each block were adjusted for each other using backward selection method. Variables that were significant at the 5% level were retained in the analysis. The significance level established for all analyses was 5% ($P \leq .05$). All analyses were performed using the Predictive Analytics Software 21.0 (PASW Statistics), formerly known as SPSS.

RESULTS

Initially, 139 eligible adults with CL/P registered at the referral center for patients with congenital malformations were contacted and invited. Of them, 114 agreed to participate in the study (response rate = 82%). Eighteen individuals were excluded because they were under current repairing treatment for oral clefts ($n = 4$), had facial disfigurement ($n = 1$) and had associated syndromes or neurological deficits ($n = 13$). The final sample consisted of 96 adults, 15 with CL±A, 22 with CP and 59 with CLP.

The description of the sample according to CL/P groups is summarized in Table 1. Participants had ten years of schooling on average and 50% of the sample reported family income greater than two minimum Brazilian wages and 71.9% of the sample were single. The mean age of the participants was 29.4 years ($SD = 9.1$), ranging from 18 to 63 years (median = 28). Of the participants, 62.5% were females. The mean score of social support and social network scales were 79.3 ($SD = 17.9$) and 2.4 ($SD = 3.7$), respectively. Most of the subjects did not smoke (95.8%), did not consume alcohol (63.5%) and did not do physical activity in

the last week (74.0%). Overall, 75% of the sample received additional procedures to primary plastic surgery. Three individuals with CP had never undergone any treatment. Of the 59 patients with CLP, five did not undergo to palatoplasty, five had the palate fully open despite having undergone the surgery and 11 had palatal fistulas. Eight subjects with bilateral cleft underwent to premaxilla resection. A third of those with clefts involving the palate had never been assessed by a speech therapist and 59% of the patients had never been submitted to orthodontic treatment. Of the 39 participants who were candidates for orthognathic surgery, only two received the procedure. In addition, 14 of the 71 patients who were candidates for alveolar bone graft were submitted to surgery. Thirty-seven percent of the individuals with cleft lip with or without cleft palate (CL±P) were submitted to rhinoplasty. The mean age of first lip repair and first palatoplasty was 4.1 and 10.9 years, respectively. Almost seventeen percent of participants had at least one chronic disease. The mean of DMFT, decayed teeth, missing teeth and DAI were 12.7, 1.2, 5.3 and 37.1, respectively. Satisfaction with CL/P treatment was reported by 63.4% of the sample, ranging from 54.2% (CP) to 86.6% (CL±A). Nevertheless, 78.1% informed they would need additional treatment, usually related to “speech”, “nose” and “teeth”. Significant differences were found for the ‘aesthetic appearance of nasolabial region’ and ‘satisfaction with facial appearance and function’ among the groups of cleft types.

The OIDP mean score was 19.6 (median = 12.3, range 0 to 78) and 76% of the sample reported at least one oral impact on daily performances (OIDP \geq 1): CL±A 53%, CP 73%, and CLP 83%. Fifty-three percent of the individuals reported impact on “smiling” and “speaking”. The highest and lowest scores were on “smiling” and “relaxing” items, respectively. The SF-36 mean score was 76.9 (median = 80.3, range 23.69 to 98.38). The most affected dimensions were “role limitations due to physical health” and “energy/fatigue”. OIDP and SF-36 total scores, OIDP performances, and SF-36 dimension scores did not differ

statistically between CL/P groups (Table 2). However, SF-36 total score and “mental health” dimension of SF-36 showed borderline statistical difference.

HRQoL and OHRQoL instruments demonstrated appropriate psychometric properties, suggesting the obtained QoL measures reflected the underlying constructs. OIDP and SF-36 Cronbach’s α were 0.818 and 0.837, respectively. The item-total correlation for OIDP varied between 0.498 (relaxing/sleeping) and 0.852 (emotional state), while the domain-total correlation for SF-36 ranged from 0.672 (role limitations due to emotional problems) to 0.858 (mental health). All items of the OIDP and SF-36 questionnaires were maintained in the regression analyses.

The unadjusted associations between independent variables and HRQoL and OHRQoL scores are presented in Table 3. There was statistically significant association of education, family income, age, sex, social support, social network of friends, physical activity, type of oral clefts, DMFT index and number of missing teeth with OHRQoL and HRQoL. Chronic diseases were also associated with worse HRQoL. Smoking, CL/P protocol compliance, number of decayed teeth and malocclusion increased the likelihood of poor OHRQoL.

In the multivariable analysis, five models tested the association of independent variables with HRQoL and OHRQoL scores. Poisson regression models for HRQoL (SF-36 scores) are shown in Table 4. In Model 1, greater education and high family income were associated with better HRQoL. Female sex was associated with low scores of HRQoL in Model 2 after adjustment for socioeconomic factors. Social ties and physical activity were added in Model 3 and Model 4, respectively. High social support scores were related to better HRQoL. In the final model (Model 5), the association of high social support and non-CP oral clefts with better HRQoL remained statistically significant. Low family income, female sex, chronic diseases and caries experience remained inversely associated with better HRQoL.

Table 5 shows the results from adjusted Poisson regression analysis on the association between independent variables and OHRQoL scores. Adults with lower education and low family income have poor OHRQoL score in Model 1. Females were more likely to have poor OHRQoL in Model 2. Lower education, low income, and female sex predicted worst OHRQoL in Model 3. These variables and social ties were associated with poor OHRQoL in Model 4. Lower education, low income, female sex, low social support, low social network, smoking, type of oral cleft, number of decayed teeth and number of missing teeth increased the mean of OHRQoL scores in the final model (Model 5).

DISCUSSION

The present study examined the role of sociodemographic factors, social ties, behavioral and biological (chronic diseases and oral clinical status) characteristics on HRQoL and OHRQoL in adults with non-syndromic CL/P using a theoretical framework. Structural and intermediary determinants are important factors associated with HRQoL and OHRQoL in adults with orofacial clefts. Overall, family income, sex, social support, type of oral cleft and dental caries were relevant aspects related to HRQoL and OHRQoL outcomes. Chronic conditions and smoking were also related to HRQoL and OHRQoL, respectively.

Several studies have demonstrated that demographic and socioeconomic characteristics are associated with the HRQoL and OHRQoL in individuals with CL/P (Marcusson et al., 2001; Damiano et al., 2007; Kramer et al., 2009; Mani et al., 2010; Broder et al., 2012; Reddy et al., 2012; Collet et al., 2013; Dak-Albab and Dashash, 2013). Our findings on the relationship of family income with HRQoL and OHRQoL are in accordance with previous studies in adults with and without CL/P (Trentini et al., 2011; Damiano et al., 2007; Dak-Albab and Dashash, 2013). Education was also directly associated with OHRQoL. A recent systematic review has suggested that children from families with high income and greater parental education have better OHRQoL (Kumar et al., 2014). A possible explanation

is the fact that people from more affluent social groups live in health-promoting environments and adopt healthier behaviors that influence health. The role of socioeconomic inequalities in explaining differences in QoL in patients with CL/P is therefore suggested in this study and reiterates the importance of the social determinants of health in this population group. Females reported higher impact on HRQoL and OHRQoL than males, which is also consistent with previous studies in individuals with CL/P (Marcusson et al., 2001; Mani et al., 2010; Broder et al., 2014b). Female patients with cleft deformities were less satisfied with their facial appearance and had poorer HRQoL than men (Marcusson et al., 2001; Mani et al., 2010).

Social support was associated with HRQoL and OHRQoL outcomes. Poor social relationships have been considered a risk factor to health (Berkman and Syme, 1979). Weak social ties may also result in limitations imposed by health problems that negatively influence the relationships between individuals, their friends and family members (Andrade and Vaitsman, 2002). Low social support has been associated with inadequate coping skills that in turn undermine healthy behaviors and exacerbate poor lifestyles favoring the occurrence of diseases (Brennan and Spencer, 2012). The high proportion of single adults with CL/P in this study is in agreement with previous findings (Ramstad et al., 1995). The number and complexity of surgeries, speech and hearing impairments, and concerns about facial appearance in individuals with CL/P resulted in a significant impact on self-rated health and social relations (Thompson and Kent, 2001; Sank et al., 2003; Sinko et al., 2005; Kramer et al., 2009). Individuals with CL/P are more prone to social isolation and negative interactions with their peers during childhood and adolescence (Hunt et al., 2005; Murray et al., 2010).

Dental caries experience was associated with worse HRQoL whereas the number of decayed teeth and the number of missing teeth were related to poor OHRQoL. The impact of dental caries, periodontal disease and malocclusion on OHRQoL was reported in studies involving subjects without CL/P (Do and Spencer, 2008; Gomes et al., 2014). The influence

of oral conditions on OHRQoL may be more relevant to individuals with CL/P as they seem to be more susceptible to oral diseases (Antonarakis et al., 2013).

Previous studies have shown that deficiencies in complying with the CL/P treatment protocol and poor satisfaction with CL/P treatment influence QoL (Brattström et al., 2005; Sinko et al., 2005; Mølsted et al., 2005; Munz et al., 2011). In this study, although the majority of the participants received further procedures in addition to primary plastic surgery, the management of CL/P was inadequate since primary plastic surgeries were conducted at late ages and clinical interventions were not integrated. In addition, few participants who were eligible for surgeries related to CL/P received the procedures, which might be related to low treatment compliance, socioeconomic disparities and healthcare inequalities. Although these deficiencies might negatively impact on QoL (Broder et al., 2014a), this was not observed in the present study. The heterogeneity in compliance with the CL/P treatment protocol may explain the lack of association between CL/P protocol compliance and QoL measures. Many individuals who needed additional treatment indicated that they were satisfied with the treatment received, although almost 80% of the sample wished further treatment and reported specific concerns about their speech, nasal cavity and the appearance of their teeth. Evidence shows that these patients may report positive subjective health after treatment despite the distress caused by the high burden of healthcare and dissatisfaction with the treatment received (Sinko et al., 2005; Oosterkamp et al., 2007; Kramer et al., 2009).

Chronic diseases were associated with HRQoL. Although no previous study has evaluated the association between chronic diseases and subjective health measures in individuals with CL/P, previous research involving individuals without CL/P have reported the relationship between chronic conditions and poor QoL (Al-Naggar et al., 2011; Vadstrup et al., 2011). In this study, smoking was associated with poor OHRQoL. The link between smoking, the occurrence of diseases and poor QoL is well established (Michelson et al., 2001; Mitra et al., 2004).

The percentage of individuals with CL/P who reported at least one impact of oral health on the daily activities ($OIDP \geq 1$) was higher compared to the general population in Brazil (Brazil, 2012). The greatest impact on 'smiling' and 'speaking' oral performances in this study reflects the aesthetic and functional impairments related to CL/P that may remain after treatment. The most affected performance influenced by oral health in individuals without CL/P was 'eating and enjoying the food' (Brazil, 2012; Yusof et al., 2013; Mohebi et al., 2014). In the studied sample, HRQoL was poorer than previous studies in adults with CL/P (Sinko et al., 2005; Oosterkamp et al., 2007; Mani et al., 2010), although it was similar to Foo and coworker's study (Foo et al., 2012). The socioeconomic and treatment-related characteristics differences between the studied sample and sample from previous studies may explain the discrepancies between their findings.

The present study has some limitations to be addressed. The cross-sectional design restricts causal inferences. The small sample size must be considered when interpreting the results since it influences the power of the study and the precision of the estimates. It is unknown the level of comprehensiveness and updating of the records of the referral center and maybe not all eligible adults with CL/P living in the city were invited. This might have influenced the sample size and randomness of the studied sample, which in turn could also have influenced the reported associations between the independent variables and QoL outcomes. It is expected that adults with CL/P who were not registered at the referral center, those who could not be reached by telephone or were not found during home visits were from socially deprived groups and have worse health status (Broder et al., 2012; Kumar et al., 2014; Garcia-Subirats et al., 2014). In order to identify adults with CL/P who could not be contacted, participants were asked whether they knew other patients with oral clefts. However, this strategy proved to be of little effect. The capacity to provide repairing treatment for CL/P at the referral center where the study was conducted implies that the present findings should not be generalized to adults with CL/P assisted by services with

different characteristics. There could be recall bias concerning the use of self-reported information about treatment history of CL/P. However, information about previous treatments for oral clefts might be reliable since they represent significant life events. Future longitudinal studies on the relationship of structural and intermediate determinants with HRQoL and OHRQoL should be conducted according to the type of cleft. A follow-up study involving the participants of the present study is nonetheless under consideration.

Studies on factors associated with QoL in patients with CL/P are scarce, although their relevance for planning and delivering better health care is without doubt. Clinical outcomes are well-recognized treatment goals in the management of the individuals with CL/P. Nonetheless, this study supports the contemporary trends of incorporating patient-reported outcome measures (PROMS) in healthcare assessment, including on treatment evaluation of individuals with CL/P. Thus, the improvement of QoL of individuals with CL/P should be one of the main treatment goals along with the aesthetic and functional gains. The overvaluation of normative clinical measures in healthcare needs assessment neglects the relevance of QoL and other subjective health measures. Overcoming the biomedical model by incorporating subjective measures to inform health care practices and evaluate treatment outcomes can improve the interventions for individuals with CL/P. In addition, the results of this study provide evidence to support multisectoral approaches to the improvement of health related outcomes including the development of health promotion actions that are necessary to tackle the broader socioeconomic and psychosocial determinants of HRQoL and OHRQoL.

Our findings highlight the role of individual and environmental determinants of HRQoL and OHRQoL in subjects with CL/P. As a result, we can argue that there is a need to involve different professionals from the health sector as well as from other areas to improve and maintain the physical, mental and social well-being of patients with oral clefts (Bircher and Kuruvilla, 2014). There is also a clear argument that individuals with CL/P can benefit

from policies aiming to reduce socioeconomic inequalities and to decrease access to health services disparities.

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Figure 1. Theoretical model for the study of structural and intermediary determinants of HRQoL and OHRQoL in adults with oral clefts (adapted from WHO, 2010).

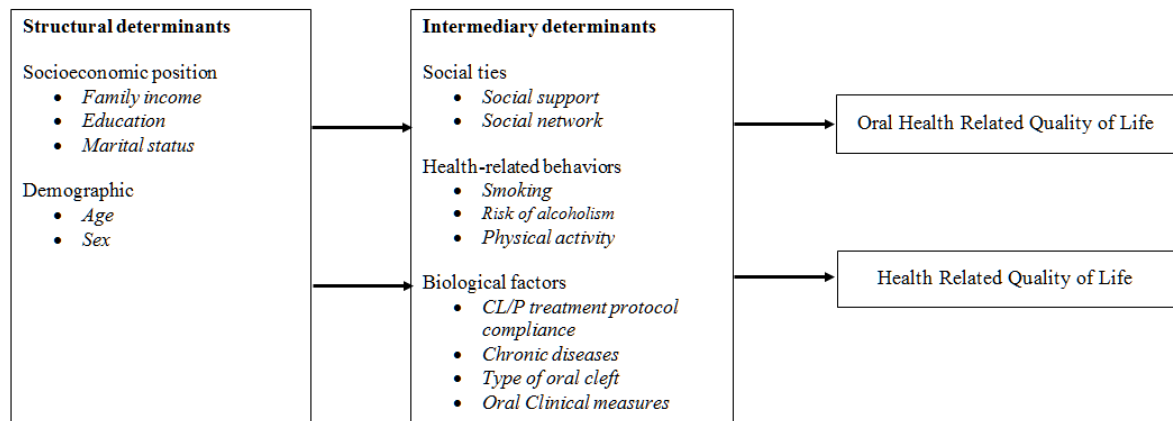


Table 1. Demographic and socioeconomic characteristics, health behaviors, chronic disease and obesity, psychosocial factors, treatment history for CL/P and clinical measures for total sample and by cleft type.

Variable	Total (n=96)	CL±A (n=15)	CP (n=22)	CLP (n=59)
Structural determinants				
Socioeconomic factors				
Education (years of schooling), mean (SD)	10.4 (3.0)	10.3 (3.2)	10.8 (2.6)	10.3 (3.2)
Family income (minimum wages), n (%)				
≤ 2 MW	48 (50.0)	8 (53.3)	9 (40.9)	31 (52.5)
> 2 MW	48 (50.0)	7 (46.7)	13 (59.1)	28 (47.5)
Marital status, n (%)				
Single	69 (71.9)	10 (66.7)	20 (90.9)	41 (69.5)
Married / living with partner	24 (25.0)	4 (26.6)	2 (9.1)	16 (27.1)
Separated / divorced	1 (1.0)	-	-	1 (1.7)
Widow	2 (2.1)	1 (6.7)	-	1 (1.7)
Demographic characteristics				
Age, mean (SD)	29.4 (9.1)	29.7 (11.2)	28.3 (7.4)	29.7 (9.2)
Sex, n (%)				
Male	36 (37.5)	4 (26.7)	6 (27.3)	26 (44.1)
Female	60 (62.5)	11 (73.3)	16 (72.7)	33 (59.9)
Intermediary determinants				
Psychosocial factors				
Social support, mean (SD)	79.3 (17.9)	84.4 (12.4)	81.6 (17.9)	77.1 (19.0)
Social network of friends, mean (SD)	2.4 (3.7)	1.9 (1.7)	1.9 (1.9)	2.7 (4.4)
Health behaviors				
Smoking, n (%)				
Yes	4 (4.2)	1 (6.7)	1 (4.5)	2 (3.4)
No	92 (95.8)	14 (93.3)	21 (95.5)	57 (96.6)
Risk of alcoholism, n (%) (n=35)				
Did not consume alcohol	61 (63.5)	10 (66.7)	16 (72.7)	35 (59.3)
No risk of alcoholism	28 (29.2)	5 (33.3)	4 (18.2)	19 (32.2)
At risk of alcoholism	7 (7.3)	-	2 (9.1)	5 (8.5)
Physical activity, n (%)				
Yes	25 (26.0)	7 (46.7)	3 (13.6)	15 (25.4)
No	71 (74.0)	8 (53.3)	19 (86.4)	44 (74.6)
Biological measures				
CL/P protocol compliance, n (%)				
No primary plastic surgery	3 (3.1)	-	3 (13.6)	-
Only primary plastic surgery	21 (21.9)	3 (20.0)	2 (9.1)	16 (27.1)
Additional procedures to primary plastic surgery	71 (75.0)	12 (80.0)	17 (77.3)	43 (72.9)
Chronic diseases, n (%)				
Yes	16 (16.7)	2 (13.3)	3 (13.6)	11 (18.6)
No	80 (83.3)	13 (86.7)	19 (86.4)	48 (81.4)
Oral clinical measures				
DMFT, mean (SD)	12.7 (7.2)	11.9 (7.3)	12.1 (5.8)	13.1 (7.8)
Number of decayed teeth, mean (SD)	1.2 (1.8)	0.9 (1.4)	1.5 (1.5)	1.1 (1.9)
Number of missing teeth, mean (SD)	5.3 (7.1)	4.1 (6.8)	4.4 (5.4)	5.9 (7.8)
DAI, mean (SD) (n=80)	37.1 (16.4)	32.7 (12.1)	34.4 (12.6)	39.5 (18.6)
CL/P related measures				
Treatment history of CL/P				
Age of the first lip repair, mean (SD) (n=70)	4.1 (6.0)	5.2 (10.1)	-	3.8 (4.4)
Age of the first palatoplasty, mean (SD) (n=69)	10.9 (10.3)	-	7.9 (7.3)	12.0 (11.0)
Craniofacial growth				

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Occlusal index (Goslon/Bauru), n (%) (n=49)				
≤ 3	29 (59.2)	-	-	29 (59.2)
≥ 4	20 (40.8)	-	-	20 (40.8)
<i>Aesthetic appearance of nasolabial region</i>				
Aesthetic appearance of nasolabial region, mean (SD) (n=50)	2.8 (0.9)	1.9 (0.6)	-	3.0 (0.8)
Nasal morphology, mean (SD)	3.10 (1.31)	2.00 (1.21)	-	3.45 (1.16)
Nasal symmetry, mean (SD)	2.76 (0.98)	1.92 (0.79)	-	3.03 (0.89)
Redness border of upper lip, mean (SD)	0.22 (0.42)	0.17 (0.39)	-	0.24 (0.43)
Nasal profile, mean (SD)	2.44 (1.26)	1.33 (0.49)	-	2.79 (1.23)
<i>Satisfaction with facial appearance and function</i>				
CEP, mean (SD)	4.3 (1.5)	5.3 (1.3)	5.0 (1.1)	3.8 (1.4)
<i>Satisfaction with CL/P treatment, n (%) (n=93)</i>				
Satisfied / very satisfied	59 (63.4)	13 (86.6)	32 (54.2)	14 (73.7)
Neither satisfied nor unsatisfied	16 (17.2)	1 (6.7)	11 (18.7)	4 (21.0)
Unsatisfied / very unsatisfied	18 (19.4)	1 (6.7)	16 (27.1)	1 (5.3)
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Table 2. Mean and standard deviation of HRQoL and OHRQoL measurements and their respective domains, in a centesimal scale (0-100).

	Total	CL±A	CP	CLP	P-value*
	Mean (DP)	Mean (DP)	Mean (DP)	Mean (DP)	
OIDP	19.6 (21.5)	14.8 (21.1)	20.1 (20.3)	20.7 (22.1)	0.310
Eating	18.9 (30.4)	12.3 (23.1)	22.4 (29.2)	19.3 (32.5)	0.767
Speaking	28.9 (37.2)	13.3 (27.7)	26.6 (35.9)	33.8 (39.1)	0.227
Cleaning teeth	15.3 (28.6)	18.9 (30.7)	18.2 (28.3)	13.3 (28.5)	0.466
Relaxing/sleeping	8.0 (20.2)	7.2 (20.7)	10.0 (23.0)	7.5 (19.2)	0.998
Smiling	36.9 (42.8)	33.3 (45.1)	35.8 (44.8)	38.2 (42.1)	0.837
Emotional status	14.9 (29.1)	8.3 (26.1)	15.3 (23.0)	16.4 (31.0)	0.359
Carrying out work or role	16.1 (31.9)	6.1 (20.6)	16.4 (32.6)	18.6 (33.9)	0.550
Social contact	18.0 (33.9)	18.7 (38.9)	16.2 (32.7)	18.4 (33.6)	0.844
SF-36	76.9 (17.8)	79.4 (15.7)	69.4 (19.2)	79.0 (17.2)	0.050
Physical Functioning	93.6 (12.4)	96.7 (6.5)	90.9 (12.3)	93.9 (13.5)	0.075
Role limitations due to physical health	81.0 (32.6)	90.0 (18.4)	68.2 (43.8)	83.5 (29.6)	0.287
Role limitations due to emotional problems	68.1 (40.4)	62.2 (48.6)	51.5 (45.7)	75.7 (34.4)	0.098
Energy/fatigue	66.4 (21.2)	72.7 (17.0)	62.5 (22.8)	66.3 (21.6)	0.361
Mental health	72.7 (20.2)	81.1 (20.4)	68.9 (17.5)	72.0 (20.9)	0.053
Social functioning	81.3 (25.8)	78.3 (21.9)	75.6 (25.7)	84.1 (26.8)	0.150
Pain	79.3 (22.8)	79.8 (22.9)	73.0 (27.8)	81.5 (20.6)	0.503
General health	72.6 (22.6)	74.3 (22.4)	65.0 (22.5)	75.0 (22.5)	0.097

*P-value refers to Kruskal-wallis test between cleft types

Table 3. Unadjusted Poisson regression for OIDP and SF-36 scores.

	OIDP			SF-36		
	β	SE	P	β	SE	P
Structural determinants						
Socioeconomic factors						
Education (years of schooling)	-0.102	0.007	< 0.001*	0.011	0.004	0.003*
Family income (Ref: >2 MW) ≤ 2 MW	0.459	0.047	<0.001*	-0.080	0.023	0.001*
Marital status (Ref: Married) Single	-0.095	0.052	0.070	-0.025	0.027	0.349
Demographic characteristics						
Age	0.015	0.002	< 0.001*	-0.004	0.001	0.001*
Sex (Ref: Male) Female	0.411	0.051	< 0.001*	-.159	0.024	<0.001*
Intermediary determinants						
Psychosocial factors						
Social support	-0.019	0.001	< 0.001*	0.004	0.001	<0.001*
Social network of friends	-0.144	0.013	< 0.001*	0.010	0.003	0.001*
Health behaviors						
Smoking (Ref: No)	0.842	0.080	< 0.001*	-0.065	0.060	0.278
Risk of alcoholism (Ref: No)	0.087	0.060	0.145	-0.050	0.048	0.303
Physical activity (Ref: Yes)	0.439	0.059	< 0.001*	-0.111	0.026	<0.001*
Biological factors						
CL/P protocol compliance (Ref: Additional procedures to primary plastic surgeries)						
No primary plastic surgery	0.309	0.052	< 0.001*	0.021	0.028	0.445
Chronic diseases (Ref: No)	0.087	0.060	0.145	-0.150	0.033	<0.001*
Type of oral cleft Ref: CP						
CLP	0.029	0.055	0.595	0.127	0.030	<0.001*
CL±	-0.303	0.082	< 0.001*	0.132	0.040	0.001*
Oral clinical measures						
DMFT index	0.028	0.003	< 0.001*	-0.007	0.002	<0.001*
Number of decayed teeth	0.136	0.010	< 0.001*	-0.003	0.007	0.674
Number of missing teeth	0.029	0.003	< 0.001*	-0.006	0.002	<0.001*
DAI	0.017	0.001	< 0.001*	-0.001	0.001	0.101

* $P < 0.05$

Table 4. Multivariate Poisson regression of socioeconomic data, health behaviors, psychosocial factors, treatment history of CL/P and clinical measures and SF-36 score.

	Model 1			Model 2			Model 3			Model 4			Model 5		
	β	SE	P	β	SE	P	β	SE	P	β	SE	P	β	SE	P
<i>Environmental characteristics</i>															
<i>Socioeconomic factors</i>															
Education	0.008	0.004	0.048*	0.006	0.005	0.210	0.005	0.004	0.227	0.005	0.004	0.194	0.000	0.005	0.959
Family income (Ref: >2 MW) ≤ 2 MW	-0.066	0.025	0.007*	-0.042	0.027	0.115	-0.014	0.025	0.593	-0.015	0.025	0.549	-0.075	0.028	0.008*
<i>Individual factors</i>															
<i>Demographic characteristics</i>															
Age				-0.003	0.002	0.064									
Sex (Ref: Male) Female				-0.137	0.025	< 0.001*	-0.142	0.025	< 0.001*	-0.132	0.026	< 0.001*	-0.103	0.026	< 0.001*
<i>Psychosocial factors</i>															
Social support							0.003	0.001	< 0.001*	0.003	0.001	< 0.001*	0.003	0.001	< 0.001*
Social network of friends							0.005	0.003	0.106						
<i>Health behaviors</i>															
Physical activity (Ref: Yes)										-0.039	0.028	0.163			
<i>Biological measures</i>															
Chronic diseases (Ref: No)													-0.146	0.034	< 0.001*
<i>Type of oral cleft Ref: CP</i>															
CLP													0.138	0.030	< 0.001*
CL±A													0.132	0.040	0.001*
<i>Oral clinical measures</i>															
DMFT index													-0.007	0.003	0.019*
Number of missing teeth													0.002	0.003	0.506

* $P < 0.05$

Table 5. Multivariate Poisson regression of socioeconomic data, health behaviors, psychosocial factors, treatment history of CL/P and clinical measures and OIDP score.

	Model 1			Model 2			Model 3			Model 4			Model 5		
	β	SE	P	β	SE	P	β	SE	P	β	SE	P	β	SE	P
Structural determinants															
Socioeconomic factors															
Education	-0.089	0.007	< 0.001*	-0.084	0.009	< 0.001*	-0.060	0.008	< 0.001*	-0.054	0.008	<0.001*	-0.081	0.013	< 0.001*
Family income (Ref: >2 MW) ≤ 2 MW	0.298	0.049	< 0.001*	0.249	0.053	< 0.001*	0.155	0.052	0.003*	0.262	0.053	< 0.001*	0.195	0.072	0.007*
Demographic characteristics															
Age				0.002	0.003	0.382									
Sex (Ref: Male) Female				0.293	0.054	< 0.001*	0.238	0.054	< 0.001*	0.255	0.056	< 0.001*	0.404	0.074	< 0.001*
Intermediary determinants															
Psychosocial factors															
Social support							-0.012	0.001	< 0.001*	-0.010	0.001	< 0.001*	-0.005	0.002	0.011*
Social network of friends							-0.070	0.012	< 0.001*	-0.078	0.013	< 0.001*	-0.073	0.015	< 0.001*
Health behaviors															
Smoking (Ref: No)										0.825	0.086	< 0.001*	0.514	0.139	<0.001*
Physical activity (Ref: Yes)										0.126	0.064	0.049*	-0.016	0.073	0.984
Biological measures															
CL/P protocol compliance (Ref: Additional procedures to primary plastic surgeries) No primary plastic surgery													0.075	0.081	0.353
Type of oral cleft Ref: CP															
CLP													0.057	0.071	0.421
CL±A													-0.276	0.102	0.007*
Oral clinical measures															
Number of decayed teeth													0.157	0.022	< 0.001*
Number of missing teeth													0.026	0.010	0.010*
DAI													0.004	0.002	0.080

* $P < 0.05$

